Aneurysm of Persistent Primitive Hypoglossal Artery Occluded with Guglielmi Detachable Coils

M. GRAND, J. NEPPER-RASMUSSEN

Department of Radiology Odense University Hospital; Denmark

Key words: persisting primitive hypoglossal artery, intracranial aneurysm, endovascular treatment, Guglielmi Detachable Coil

Summary

The persistent primitive hypoglossal artery is rare remnant of one of the four embryonal carotid-basilar anastomoses. It is present in 0.02-0.26% of all cerebral angiogram. 14 cases of PHA aneurysms have been reportede in the literature and as far as we know no case in which endovascular embolization was used as a treatment. We present a case with subarachnoid haemorrhage due to aneurysm of persistent primitive hypoglossal artery. The aneurysm was successfully occluded with Guglielmi detachable coils. The SAH resolved and recovery was uneventful.

Introduction

In early embryonic stage at the 4 mm stage several anastomtic channels run from the ventral primitive internal Carotid arteries and dorsal aorta to the paired longitudinal neural arteries. The most rostral of these are the trigeminal, otic and hypoglossal arteries. These arteries regress synchronously with the development of the posterior communicating and basilar arteries and have disappeared normally by the 14 mm stage of the embryo [1].

The trigeminal artery is the most common of the primitive carotid-basilar anastomoses that persist into adulthood and persistent primitive hypoglossal artery (PHA) is the second most frequent one. PHA was first demonstrated in 1950 by angiography. Since then, several cases of an aneurysm of persistent primitive hypoglossal artery (PHAA) have been reported. Review of the literature documents 15 cases of PHAA including our case and as far as we know there has been no case in the literature in which a PHAA has been treated with endovascular embolization [2].

Case Report

A 38-year-old woman experienced a sudden occipital headache followed by unconsciousness. Plain CT performed on admission showed subarachnoid haemorrhage (SAH) and mild degree of hydrocephalus. Following CT-angiography performed two days later aroused suspicion of an aneurysm on the right PICA. The patient was referred to our hospital, which at that time was the only GDC center in Denmark. A cerebral angiography disclosed a PHA on the right side communicating with internal carotid and vertebral artery and an aneurysm on the PHA.

The PHAA was located at the junction between the PHA and the vertebral artery (VA), and with the tip pointing caudally. It had a wide neck and measured 2.5 x 4.5 mm. The right VA was persistent but hypoplastic (figure 1).

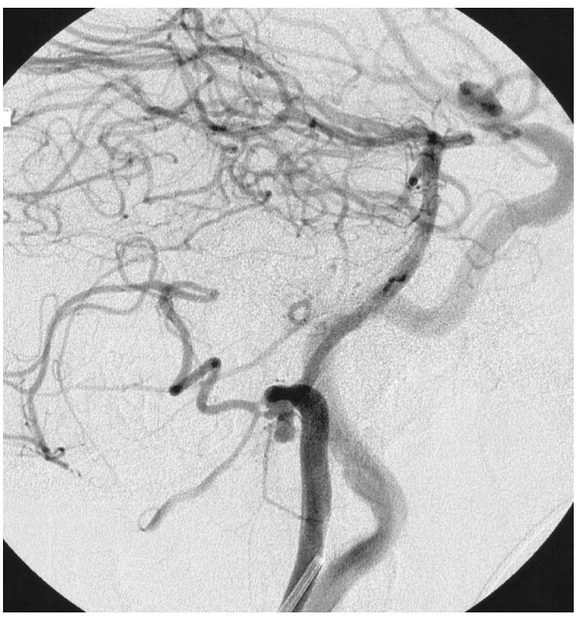

Figure 1 Pre-embolization angiogram of the right ICA demonstrates hypoglossal artery aneurysm. The right VA is hypoplastic.

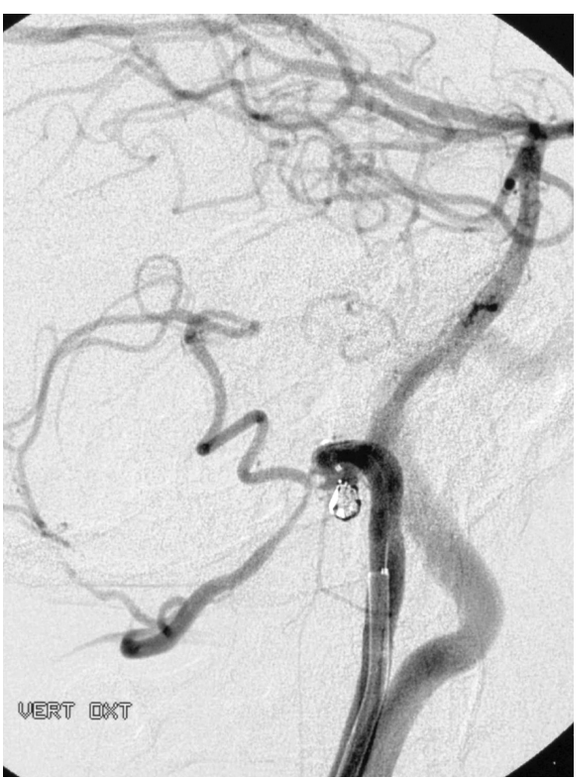

Figure 2 Post-embolization angiogram after occlusion of the aneurysm. Two GDC are successfully introduced into the aneurysm.

The patient was a candidate for endovascular treatment with GDC, which is the first choice for treatment of aneurysms in our hospital. By a femoral approach a guiding catheter was placed into the right internal carotid artery. A microcatheter was introduced into the aneurysm sac via the PHA, and two GDC were placed in the aneurysm giving a good occlusion of the sac and normal flow in the PHA as well as the VA (figure 2).

The patient's post course was uneventful, and she was able to resume to her normal life. A follow up postoperative angiogram two years later showed a well occluded aneurysm without compaction of the coils and persisting circulation (figure 3).

Discussion

The carotid-basilar anastomoses are well known developmental vascular anomalies, and of these anomalies a persistent trigeminal artery is the most common one. Aneurysms of this artery have been demonstrated several times, and also endovascular treatment has been described [3]. PHA is much rarer and is demonstrated in about 0.02 to 0.26% of cerebral angiograms [4,5]. PHA is functionally a single artery irrigating the brainstem and cerebellum.

There are four criteria used for detection of the hypoglossal artery [6].

1) The artery must arise from the cervical part of the internal carotid artery above the level of C3.

2) The artery must enter the posterior fossa via hypoglossal canal.

3) The basilar artery only fills beyond the point at which the hypoglossal artery enters.

4) The posterior communicating artery on the same side is absent or hypoplastic.

A review of the literature revealed a total of 14 cases of PHAA. Eight patients were female and the age of the patients ranged from 10 to 62. The PHAA was on the right side in eight cases, on the left side in five cases and both sides in one case. Of eleven patients treated surgically, eight had successful aneurismal clipping and good postoperative course. The aneurysm in the remaining three patients was incompletely clipped and in two patients it was wrapped with dental cotton because of the size of the aneurysm [2].

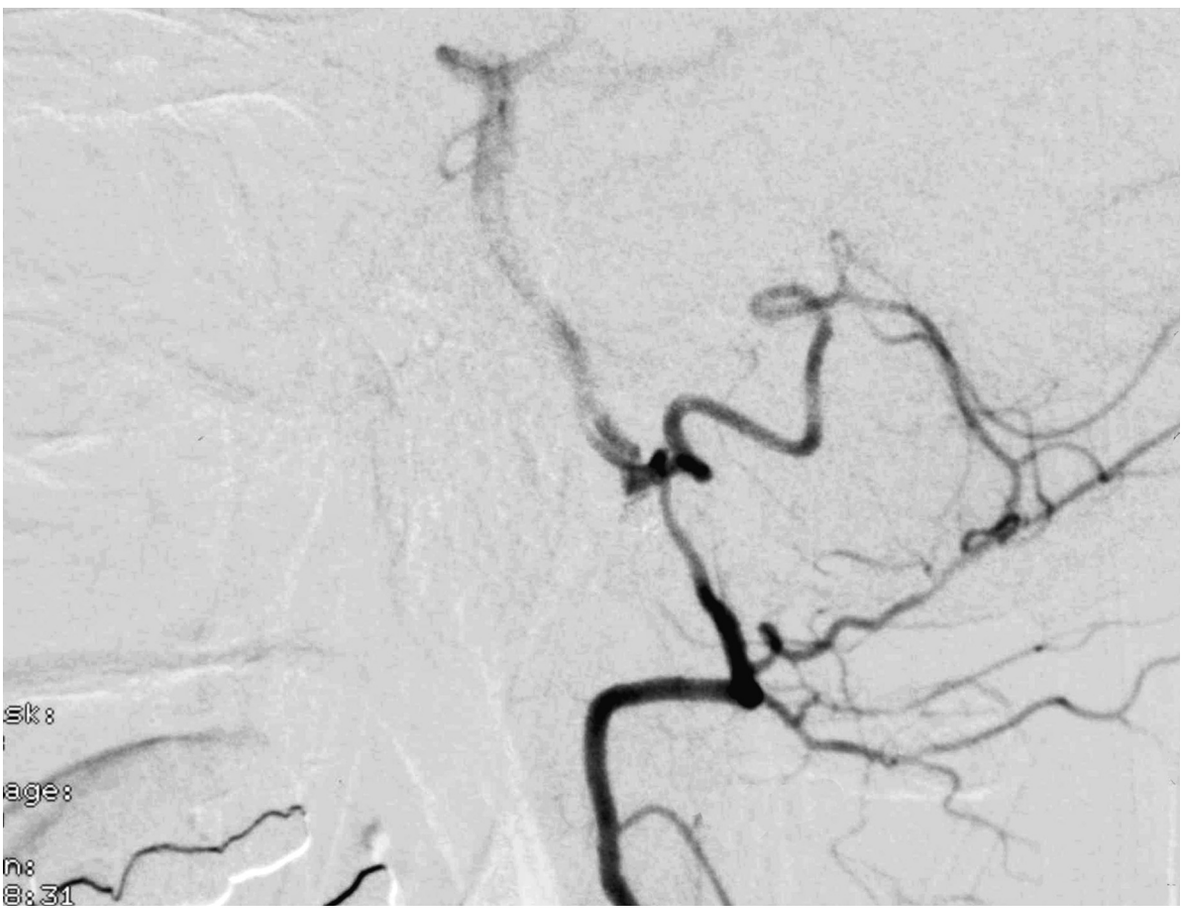

Figure 3 Follow-up angiogram two years later after occlusion shows a well occluded aneurysm without compaction of the GDC.

The ultimate goal of treatment is to exclude the aneurismal sac from the intracranial circulation while preserving the parent artery. Treatments of intracranial aneurysms have long been the domain of the neurosurgeons, but since 1991 neuroradiologists have been using GDC technique to treat increasing number of patients with intracranial aneurysms [7]. Endovascular treatment is now accepted as an alternative to surgical clipping in most cases of intracranial aneurysms, especially for aneurysms in the posterior circulation, where surgery is more difficult and complicated [8]. In our case the best way to reach the aneurysm was from the internal carotid artery through the PHA, because the vertebral artery, even it was open, was hypoplastic.

Many factors have influence on obliteration rate, but the most important one is the ratio of the neck of the aneurysm to the dome. The aneurysms with wide necks are less amenable to GDC therapy than those with narrow necks, because with a wide-necked aneurysm the coils tend to compact into the body and dome of the aneurysm, resulting in a remnant aneurysm and incomplete treatment. The access to a proximal located aneurysm as this PHAA is easy, giving a low procedural risk. The long time follow up has shown a low risk of re-bleeding [9,10]. In our case there were no difficulties in placing the coils, and follow up revealed a well occluded aneurysm. This case emphases that endovascular occlusion of a PHAA is an effective and safe option.

References

1 Anderson M: Persistent primitive hypoglossal artery with basilar aneurysm. J Neurol 213: 377-381, 1976.
2 Yokota N, Yokoyama T et Al: Aneurysm of persistent primitive hypoglossal artery. Br J Neurosurg 13: 608-610, 1999.
3 Rasmussen JN, Bjerre P, Andersen P: Aneurysm On a Persisting Primitive Trigeminal Artery Occluded With GDC. Interventional Neuroradiology 4: 91-93, 1998.
4 Kanai H, Nagai H et Al: A large aneurysm of the persistent primitive hypoglossal artery. Neurosurgery 30: 794-797, 1992.
5 Vlychou M, Georganas M et Al: Angiographic findings and clinical implications of persistent primitive hypoglossal artery. BMC Med Imaging 23; 3 (1): 2, 2003.
6 Lasjaunias P, Berenstein A: Surgical Neuroangiography. Vol 1, Chapt 3. Springer-Verlag. Berlin Heidelberg 1987.
7 Schievink WI: Intracranial aneurysms: The New England Journal of Medicine. Vol 336 (1): 28-40, 1997.
8 International Subarachnoid Aneurysm Trial (ISAT) Collaborative Group: International Subarachnoid Aneurysm Trial (ISAT) of neurosurgical clipping versus endovascular coiling in 2143 patients with ruptured intracranial aneurysms: a randomised trial. The Lancet 360: 1267-1274, 2002.
9 Fuse A, Rodesch G et Al: Endovascular Management of Intradural Berry Aneurysms. Interventional Neuroradiology 6: 27-39, 2000.
10 Murayama Y, Nien YL et Al: Guglielmi Detachable Coil embolization of cerebral aneurysms: 11 years' experience. J Neurosurg 98: 959-966, 2003.

Mansour Grand, M.D.
Sdr. Boulevard 164, lejl. 2
DK-5000 Odense C
Denmark
E-mail: grand@dadlnet.dk